CASE REPORT

Musculoskeletal syndrome treated with global postural re-education in double-redo lung transplantation: a case report with an 8-month follow-up

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SUMMARY

Postoperative pain and persisting fatigue represent critical concerns for patients receiving lung transplantation. The purpose of this study was to illustrate the trajectory of symptoms in a patient who presented with a post-transplant musculoskeletal syndrome after double redo-lung transplantation and attended therapeutic sessions of global postural re-education during the symptomatic phase.

A 32-year-old woman with interstitial lung disease underwent double lung transplantation. At 23 months, functional parameters deteriorated, and the patient was placed on the active list for a second double-lung transplantation. Twenty months after re-transplantation, the patient reported continuous thoracic-lumbar musculoskeletal pain exacerbated by moving or performing the standard motor activities.

Lower body flexibility improved during the observation period changed from -10 cm to 0 cm at the Chair Sit-and-Reach Test. Leg strength improved as well, and the patient was able to perform more repetitions at the Squat Test, improving from 14 to 39. Pain intensity changed from 7 to 4 on a numerical rating scale.

We observed that outcomes strictly related to treatment, with lower body flexibility, pain intensity, and physical function improving over time. As a result global postural re-education proved to be effective in this patient.

Key words: *Dyspnea, exercise, global postural re-education, physiotherapy, lung transplantation.*

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INTRODUCTION

Postoperative pain and persisting fatigue represent critical concerns for patients receiving lung transplantation (LTx) (1-3). Indeed, chronic pain in LTx recipients is common during the first five years, with women experiencing more intense pain than men (2). In this regard, a cohort of LTx recipients described the rehabilitative treatment as being a crucial intervention in improving their mental and physical health; therefore expectations of outcomes from their rehabilitation are high in these patients (4).

The post-transplant musculoskeletal syndrome is an umbrella term that can be used to describe symptoms affecting the musculoskeletal system associated with pain in patients receiving LTx. Postoperative pain has been found to be a common symptom that can last months to years, affecting women more than men and having a prevalence ranging from 49% to 59% (1, 5-7). On the other hand, a cyclosporine immunosuppressive regimen can be associated with muscle disorders such as myalgia, cramps, and muscle weakness (8) that can worsen in presence of pre-existing musculoskeletal alterations (*i.e.* spinal alterations, neu-

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ropathies, myopathies, joint disorders) or physical limitations caused by the underlying diseases (*i.e*. muscle deconditioning, exercise limitation, prolonged bed rest) or re-transplantation.

Although an extensive body of literature describing the rehabilitative treatment for patients receiving LTx is available, at the time of writing no published studies describe the use of global postural re-education (GPR) in such a population. GPR is a manual technique that has been already described for the treatment of various musculoskeletal conditions, such as low back pain, ankylosing spondylitis, patellofemoral pain syndrome, neck pain, temporomandibular disorders, lumbar and cervical disc herniation (9-16). In a systematic review of patients with pregnancy-related low back pain, chronic low back pain, and ankylosing spondylitis, GPR was effective in reducing pain and improving disability and functional capacity, although it was not superior to other treatments for neck pain, temporomandibular disorders, and patellofemoral pain syndrome, chronic low back pain, and ankylosing spondylitis (9). Another study confirmed that GPR was more effective than manual therapy in a cohort of 94 patients with chronic non-specific neck pain who obtained improvements in pain and disability (15). In another study conducted among 24 patients (71% women) with herniated lumbar disc, GPR contributed to reduce pain and improve function even in those who had symptoms for more than six months (10). To the same extent, another case reported improvements in pain and range of motion in a patient with a cervical disc herniation (16). Since the first review on GPR dating back to 2007 (17), several studies have been published and another review has determined that GPR has increasingly been used for the treatment of different musculoskeletal conditions, as previously highlighted (9).

The purpose of the current study was to illustrate the bahaviour of symptoms in a patient who presented with a post-transplant musculoskeletal syndrome characterised by thoracic-lumbar back pain after a double redo-LTx and attended GPR therapeutic sessions during the symptomatic phase.

■ CASE REPORT

The patient provided written informed consent. A 32-year-old woman with interstitial lung disease underwent double lung transplantation (DLTx). At the time of first transplantation, she was not involved in any sporting activity and the body mass index (BMI) was 20.3 (kg/m^2). The postoperative course was uneventful, and she was discharged home on postoperative day (POD) 28 with complete autonomy. The first postoperative spirometric evaluation performed twenty days after DLTx showed a moderate mixed (restrictive/obstructive) respiratory deficit with forced vital capacity (FVC) =1.94 L (55% predicted), forced expiratory volume in 1 second (FEV_1) =1.56 L (51% predicted), forced expiratory flow (FEF_{25-75}) =1.51 L/s (39% predicted), and FEV_1/FVC =80%. Fourteen months after DLTx, the patient clinical conditions worsened due to the development of bronchiolitis obliterans syndrome (BOS) requiring O_2 (4-6 L/min.) supplementation during motor activities such as personal hygiene (bathing), walking, climbing a flight of stairs, and performing general daily activities. In this phase, the patient continued to walk as much as possible.

At 23 months, functional parameters deteriorated and she was placed on the active list for a second DLTx. Spirometric evaluation confirmed severe mixed (restrictive/obstructive) ventilatory deficit with FVC =1.14 L (33% predicted), FEV_1 =0.52 L (17% predicted), FEF_{25-75} =1.62 L/s, and FEV_1/FVC =46%. Twenty-eight months after the first transplantation, the patient underwent a second DLTx because of respiratory insufficiency in BOS, and she was discharged home on POD 22 without the need for O_2 support. Before the second DLTx, the patient BMI was 22.2. Twenty months after re-transplantation, the patient reported continuous thoracic-lumbar musculoskeletal pain exacerbated by movement or execution of the normal motor activity. Moreover, the patient body posture was characterised by the presence of a kyphotic habitus together with internal rotation of the shoulders.

Postoperative rehabilitation was provided during the hospital stay, but the patient did not attend a long-term rehabilitation programme; she exercised at home by cycling and walking in autonomy. In this regard, we would highlight that a long-term postoperative programme should be implemented as a default condition promoting the development of a rehabilitative culture when it is not already present. To the same extent, patients should be encouraged in participating in long-term rehabilitative activities. In the last decade, tremendous improvements have been achieved in making rehabilitation available after LTx amid transplantation centres worldwide; however, additional efforts should be made to promote patients' adherence.

Magnetic resonance imaging examination did not reveal vertebral structural alterations; the size of the spinal canal was normal. Posterior vertebral walls were normally aligned, and the height of vertebral bodies was regular; intervertebral discs were of average amplitude and showed no signs of degeneration.

Osteopenia, resulting from the evolution of the underlying disease and postoperative immunosuppressive treatment, can be frequently encountered in solid organ transplantation, including LTx (18); dual-X-ray absorptiometry examination confirmed an osteopenic value (lumbar spine T-score –2.2/–2.4) in the present case.

Spirometric evaluation performed before treatment showed no significant alterations of the pulmonary function: FVC =2.66 L (79% predicted), FEV_1 =2.31 L (79% predicted), FEF_{25-75} =3.27 L/s (87% predicted), and FEV_1/FVC =86.7%. Lung function tests were executed in accordance with the ERS/ATS guidelines (19).

Dyspnea perception was rated 20 on the 0-100 Barthel Index-dyspnea (BI-d) grading score, where 0 represents no dyspnea and 100 is the maximum level of dyspnea perceived (20). The BI-d questionnaire is composed of 11 items: grooming, bathing, feeding, toilet use, using stairs, dressing, bowel movement, bladder discharge, mobility, wheelchair use, and transfers (bed to chair and back). This questionnaire can be answered choosing from five possible options namely no dyspnea, slight, moderate, severe, and extremely severe dyspnea. The sum of all answers for each item returns the final score, with higher scores representing worsening of symptoms (20).

Pain intensity was rated 7 on a 0-10 numerical rating scale (NRS), where 0 represents the absence of pain and 10 is the maximum pain perceived (21-23).

Lower body flexibility was evaluated with the Chair Sit-and-Reach (CSR) Test (24): the patient is in a sitting position at the edge of a chair (43 cm height). One leg is extended with the ankle flexed at 90°, and the other leg is with the knee flexed and the foot on the floor. The subject is asked to slowly reach - with the hands overlapping - the tip of the toe; the distance measured (in cm) between the fingertips and the toe represents the test result.

Leg strength was evaluated by the number of repetitions made by the patient at the Squat Test, which is administered to evaluate the strength of lower body muscles (quadriceps, hamstrings, gluteus). The subject is asked to squat down and touch the chair behind them repeating this movement until they get fatigued (25, 26). Results are determined by the number of repetitions performed before getting fatigued.

At baseline, the patient was undergoing immunosuppressive therapy with tacrolimus (4.5 mg/day) and mycophenolate mofetil (1 g/day) in association with prednisone (5 mg/day). Oral supplementation of calcium carbonate and vitamin D 600 mg/400 UI/day plus cholecalciferol 25,000 UI once a month were also administered. At the onset of symptoms, the patient was prescribed paracetamol 2000mg/day for four weeks and tramadol (max 100 mg/day) for two weeks.

Considering the characteristics of symptoms and signs, a post-transplant musculoskeletal syndrome primarily characterised by thoracic-lumbar back pain was diagnosed (1-3), so the patient was referred for physiotherapy. When treatment commenced, the patient was able to perform daily activities and walk without O_2 supplementation with a peripheral oxygen saturation of 96% in room air. During the treatment, the patient was monitored with a pulse oximeter checking O_2 saturation ≥95%. The treatment consisted of GPR, which is a manual method created by P. Souchard in 1981 to rebalance altered postural patterns. For the present case, the rationale was supported by the hypothesis that the musculoskeletal system influences body shapes and can impair functions (27). The purpose of GPR is to restore the patient's lost function, ensuring a reduction in pain and an improvement of mobility. Daily practice shows us that, in presence of pain and function reduction, there are always changes in the position of the single joint or of the body segment of which it is part: recovering function improves posture. One of the concepts of GPR is the role of eccentric contraction; the isometric contraction in the most eccentric position allows to reduce the neuromuscular tone. The joints and body segments deviate in the direction of muscle stiffness, the eccentric isometric contraction allows to reduce pain and decrease muscle stiffness, facilitating joint mobility at the same time. Additional considerations should be made about the role of expiration during postures; the spinal muscles play a role as accessory inspiratory muscles. Deep and slow breathing allows to overcome the passive resistance of the spinal muscles and bring them into lengthening. Expiration facilitates the distension of the inspiratory muscles, making them more flexible and therefore more mobile over time. Deep and slow breathing, as a component of various relaxation techniques, is a complementary approach in the treatment of chronic pain syndromes. It has been confirmed that the way of breathing influences pain processing, and it represents an essential feature in the modulation of sympathetic arousal and pain perception (28).

In the case described here, 10 GPR sessions were provided over three months, and

Figure 1 - Treatment positions. For illustrative purposes, a model rather than a patient has been used to demonstrate the treatment postures. A) The patient lies supine: progression towards hips extension and upper limbs adduction. B) The patient lies supine: progression towards hip extension and upper limb abduction. C) The patient is standing against the wall: progression towards the adduction of the upper limbs. Stress on self-stretching of the spine.

Table I - Changes of clinical variables over the observation period.

Variables	Baseline	Follow-up		
		1th (15-days)	2nd (5-months)	3rd (8-months)
Dyspnea BI-d (score)	20	21	34	45
Pain NRS (score)	7	5	4	4
Lower body flexibility CSR (cm)	10	10	13	0
Leg strength ST (no. of repetitions)	14	17	32	39

BI-d, Barthel Index dyspnea; NRS, numerical rating scale; CSR, Chair Sit-and-Reach Test; ST, Squat Test.

after a 45-day pause, other 8 GPR treatments were carried out in the following seven months. Overall, the patient attended 18 GPR sessions over 11 months. The first follow-up was planned at 15 days after the first ten sessions were completed. Then, additional follow-up visits were scheduled at 5 and 8 months (Table I), while the patient attended the remaining sessions of the whole cycle. GPR sessions lasted 1 hour each and comprised three treatment postures:

1) supine with adducted arms (Figure 1A);
2) supine with open arms (Figure 1B);
3) standing against the wall (Figure 1C).

The first and the second treatment positions are indicated for the treatment of morphological alterations of the vertebral column (27). The third posture is instead a compelling choice for the correction of shoulder girdle alterations (*i.e.*, shoulder internal rotation). A detailed description of each treatment posture has already been provided in detail elsewhere (27). The treatment postures were associated during each session considering the patient's fatigue and her understanding of the exercise. The treatment position upright against the wall was performed at the end of each session, having the patient accustomed to the technique. There were no events associated with the treatment.

Lower body flexibility improved during the observation period changing from -10 cm to 0 cm. In the same way, leg strength substantially improved as the patient was able to perform more repetitions at the Squat Test executed at eight months, improving from 14 to 39 (Table I). Finally, pain intensity decreased from 7 to 4, improving by 43%. These data, taken at baseline and last follow-up, are supporting clinically significant improvements. Conversely, dyspnea intensity deteriorated to some extent during the observation period, as illustrated in Table I. Feeding, use of the toilet, dressing/undressing, and walking/mobility were the domains presenting primary deterioration, increasing from 0 to 5, 0 to 5, 2 to 5, 3 to 12 respectively.

Pulmonary function tests did not change significantly during the 8-month study period if compared with data at baseline, confirming a stable clinical frame. In this regard, FVC ranged between 2.45 to 2.74 L (72-81% predicted, respectively); FEV_1 between 2.19 to 2.38 L (75-81% predicted, respectively); FEF_{25-75} between 2.68 to 3.45 L/s (72-92% predicted, respectively); and FEV_1/FVC between 82.22 to 87.96%.

DISCUSSION AND CONCLUSIONS

It is well known that patients undergoing LTx are at high risk for respiratory infections due to the immunosuppressive regimen administered after surgery. Such a risk often prevents them from adhering to a postoperative rehabilitative programme, given the need to leave home for attending therapeutic sessions. Facing these challenges, telerehabilitation can be particularly attractive for patients receiving LTx who can exercise at home under remote supervision (29). Although appealing, telerehabilitation is not always a viable option, as shown in the current case because of the need to perform manual techniques requiring the patient's physical presence.

We observed significant improvements in those outcomes strictly related to the intervention, despite the increased intensity of dyspnea perceived during daily activities. In fact, dyspnea worsened (Table I) as might occur in patients receiving DLTx over a given period, postoperatively. Such a trend was not related to the deterioration of motor function, especially when considering static tasks (motor performance not associated with aerobic capacity). We should keep in mind that this case involves lung re-transplantation, a phenotype with a poorer prognosis if compared to primary LTx. If on the one hand, GPR was not effective at improving a natural negative trend in dyspnea perception, on the other it substantially contributed to preserving motor activities and reducing pain. In addition, we found that BI-d was perfectly suited to detect clinical changes in our case; thanks to the presence of several descriptors referring to daily activities, it reflected the most challenging tasks for the patient.

Instead, pain intensity came below the cutoff value of NRS 5 (Table I), recognized as the value above which the pain is defined as substantial (3). In this regard, our results were in accordance with those presented in a previously published study reporting improvement in pain intensity and lower body flexibility using GPR in a cohort of patients with non-specific low back pain (11). Even in our case, low body flexibility substantially improved over time (Table I) and changes were consistent at 8-month follow-up probably reflecting that the patient was accustomed to exercise. To the same extent, even the number of repetitions at the Squat Test increased substantially over the study period and were confirmed at the 8-month follow-up, as shown in Table I. Moreover, we feel GPR could be an interesting method worthy of further investigation considering its application to treat muscular and morphological alterations, even at an early postoperative stage after LTx.

Nevertheless, the results of the present study should be considered with caution because they are based on a single case and, therefore, cannot be extended to a wider context. In addition, we preferred using specific clinical tests, such as the evaluation of low body flexibility and pain to obtain objective measures instead of subjective criteria. Despite this, other limitations could be represented by the absence of specific questionnaires evaluating low back function (30), and measurement of the kyphosis. Eventually, we cannot compare our findings with other similar cases since there are no previous published studies discussing the use of GPR in patients receiving LTx.

From the current case, we observed that outcomes strictly related to treatment, such as low body flexibility, pain intensity, and physical function, improved over time so that GPR was effective for this patient.

Conflict of Interests

The authors declare no potential conflict of interests.

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